The combination of bed sharing and maternal smoking leads to a greatly increased risk of sudden unexpected death in infancy: the New Zealand SUDI Nationwide Case Control Study

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ABSTRACT

BACKGROUND: Despite a major reduction in overall infant mortality, sudden unexpected death in infancy (SUDI) continues to be of concern in New Zealand, as the rate is high by international standards, and is even higher in indigenous Māori.

AIM: To identify modifiable risk factors for SUDI.

METHODS: A three-year (1 March 2012–28 February 2015) nationwide case-control study was conducted in New Zealand.

RESULTS: There were 137 SUDI cases, giving a SUDI mortality rate of 0.76/1,000 live births. The rate for Māori was 1.41/1,000, Pacific 1.01/1,000 and non-Māori non-Pacific (predominantly European) 0.50/1,000. The parent(s) of 97% of the SUDI cases were interviewed. Six hundred and forty-nine controls were selected and 258 (40%) were interviewed. The two major risk factors for SUDI were: maternal smoking in pregnancy (adjusted OR=6.01, 95% CI=2.97, 12.15) and bed sharing (aOR=4.96, 95% CI=2.55, 9.64). There was a significant interaction (p=0.002) between bed sharing and antenatal maternal smoking. Infants exposed to both risk factors had a markedly increased risk of SUDI (aOR=32.8, 95% CI=11.2, 95.8) compared with infants not exposed to either risk factor. Infants not sharing the parental bedroom were also at increased risk of SUDI (aOR=2.77, 95% CI=1.45, 5.30). Just 21 cases over the three-year study were not exposed to smoking in pregnancy, bed sharing or front or side sleeping position.

CONCLUSIONS: This study has shown that many of the risk factors that were identified in the original New Zealand Cot Death Study (1987–1989) are still relevant today. The combination of maternal smoking in pregnancy and bed sharing is extremely hazardous for infants. Furthermore, our findings indicate that the SUDI prevention messages are still applicable today and should be reinforced. SUDI mortality could be reduced to just seven p.a. in New Zealand (approximately one in 10,000 live births).

Abbreviations	
aOR	Adjusted odds ratio
CI	Confidence interval
NIIO	National Initial Investigation Office
OR	Odds ratio
PAR	Population attributable risk
PMMRC	Perinatal and Maternal Mortality Review Committee
SIDS	Sudden infant death syndrome
SUDI	Sudden unexpected death in infancy

We previously conducted a three-year (1987–1990) case-control study examining risk factors for sudden infant death syndrome (SIDS), the New Zealand Cot Death Study.[1,2] The major risk factors identified were prone sleeping position, maternal smoking, lack of breast-feeding and bed sharing. We subsequently showed that there was an interaction between bed sharing and smoking, so that infants of mothers who smoked in pregnancy were at a much higher risk of death when bed sharing than infants of mothers who did not smoke.[3]

The study identified several new risk factors for SIDS, including the interaction between bed sharing and smoking,[3] side sleeping position,[2] postnatal depression,[4] the independent effect of smoking by the father,[5] the protective effect of pacifiers[6] and the protective effect of sleeping in same bedroom as parents.[7] We also showed that the high rate of SIDS in Māori is based largely on the high prevalence of risk factors (especially smoking and bed sharing) in the Māori population.[8]

In February 1991, the official SIDS prevention campaign began,[9] although the prevalence of prone sleeping position had started to decrease from August 1989.[10] Within 1–2 years there was a substantial reduction in SIDS (from 250 to 120 deaths p.a.) and total postneonatal mortality rates.[11]

We followed the original study with a prospective case-cohort study with data collected from 1991 to 1993. This confirmed the dramatic decrease in the prevalence on prone sleep position and demonstrated that the previously described risk factors were still important.[12]

Terminology changed from cot death (crib death in the US) to SIDS, which is unexplained infant death and is a diagnosis of exclusion. More recently Sudden Unexpected Death in Infancy (SUDI) is used because a thorough clinical history, a review of details of the circumstances of death and the autopsy may provide a contributory or causative diagnosis. Furthermore, some pathologists and coroners prefer to use the term 'undetermined' or 'unascertained' for a death previously considered to be SIDS. This change is causing diagnostic shift in the mortality data. A set of ICD-10 codes that encompasses the codes used in different countries for most SUDI cases have been proposed.[13] Use of these codes will allow for better comparisons over time and place.

The original New Zealand Cot Death Study is now more than 25 years old, and the prevalence of risk factors has changed due to the SIDS prevention programme, and this may have changed the relative importance of the risk factors at a population basis. In 2010, there was concern about SIDS mortality rates in New Zealand as mortality rates had plateaued in the previous decade and were higher than other comparable countries.[14] Furthermore, 62% of cases were in Māori (CYMRC, 2009) and over 50% occurred in a co-sleeping context.[15,16] There were knowledge gaps, including lack of information on individual SUDI cases and the estimation of the current prevalence of risk factors in the community as these previously were based on small surveys in Auckland.[17,18] Thus it was felt appropriate to reinvestigate this problem.

The aim of this study is to reduce New Zealand's high infant mortality rate,

especially in Māori, by carrying out a nationwide study to identify the modifiable risk factors for sudden unexpected death in infancy (SUDI) using a more detailed death-scene investigation protocol in collaboration with the coronial investigation of deaths across New Zealand.

Methods

A prospective national case-control study enrolled cases with deaths occurring from 1 March 2012 to 28 February 2015. The source population for this study was the whole of New Zealand. The number of live births in the years 2012–2014 was used as the denominator for the calculation of mortality rates.

Cases

The death of an infant that was referred to the coroner was potentially eligible for inclusion. The cases had to be born and domiciled in New Zealand, and be between seven days of age and the first birthday (post-perinatal age group).

SUDI cases included the following categories of death:

- Clear asphyxia deaths occurring during sleep
- Unsafe sleeping, ie, bed sharing with no direct evidence of facial occlusion, wedging, sleeping on couch or in car seat. Prone and side sleeping position were not included in this category
- Congenital anomalies, infection and other findings insufficient to explain the death
- Unascertained and
- Unexplained causes of sudden unexpected death (normal history, autopsy and scene investigation, which fulfils the usual definition of SIDS)

It excluded

- Non accidental injury, including suspected homicide and neglect, obvious accidental causes, such as road traffic crashes and concealed pregnancies
- No autopsy (parental objection)
- Perinatal asphyxia, prenatal problems and complications of prematurity
- Clearly identified cause at autopsy with prodromal symptoms and signs
- Congenital anomalies that clearly led to death

Cases could be categorised in more than one category. Note that this definition of SUDI is broader than the definition of SIDS.

All sudden, unnatural, violent or unexplained deaths have to be reported to the Coroner.[19] All infant deaths referred to the coroner were potentially cases for the study. In New Zealand, the National Initial Investigation Office (NIIO) is a single point of contact for cases to be referred to the coroner. NIIO staff were responsible for notifying the project manager (MM) that a SUDI had occurred. At times it was initially unclear to NIIO whether the death was within the scope of the study, for example, a death of an infant with a pre-existing medical condition in a bed-sharing situation. NIIO were advised to make the notification even if they were unsure the case was within scope for the study. The project manager would confirm whether the death was in scope and, if necessary, would seek advice from the lead investigator (EAM). In all cases, the SUDI liaison sought clearance from New Zealand police prior to making first contact with the family. This provided opportunity for the SUDI liaison to be informed about whether the death was considered by New Zealand police to be suspicious and to obtain contextual and relevant background information. Cases "known to the justice system" were excluded only if they met the exclusion criteria. In cases where New Zealand police were considering a case of criminal culpability for the infant's death, the SUDI liaison would maintain regular contact with the designated police officer until such time as the death was no longer considered to be suspicious or clearance was given for the SUDI liaison to contact the family. The timeframe for this varied from one day to several weeks, however, the majority of cases were cleared of suspicion within 3–5 days.

It was anticipated that autopsies would be carried out on a high proportion of cases and that these would be carried out by forensic or perinatal/paediatric pathologists. Full autopsies were conducted predominantly by forensic and paediatric pathologists following a standard protocol modified from the International SUDI Protocol to conform to cultural guidelines and New Zealand Coronial Practice. (This included measurement of body weight and dimensions, assessment of nutritional status, the

recording of the weights of all major organs and histological examination of sections from each lung, the myocardium, trachea, medulla, cerebellum and thalamus and all major organs as well as all macroscopic abnormalities. Vitreous humour biochemistry, bacterial cultures of lung and blood, virological studies for the main respiratory viruses and toxicology were also performed.)

Data collection for cases

After notification by NIIO there was an initial assessment by a specially trained investigator (SUDI liaison), which was conducted under the auspices of the coroner. This included a death scene investigation, which also included photography and doll reconstruction of the position in which the infant was placed to sleep and found dead and of the sleeping surface and bedding. A detailed research interview (with informed consent) with the caregivers was also undertaken subsequently or at the same time as the initial assessment.

Allocation of a cause of death

An expert group comprising two pathologists, two paediatricians, a public health physician and the project manager met and considered the information from the initial and research datasets and the pathology report and classified the cause of death for each case in the study. This was done independently from the certified cause of death or the cause of death determined by the coroner.

Controls

The following method was used to select controls:

1. A date of interview (nominated date) was randomly selected from all days in the three-year study (1 March 2012 to 28 February 2015).
2. The control was then randomly allocated an age at which to be interviewed to ensure that the control group had a similar age distribution to that previously described for cases.
3. The date of birth was calculated from the age and nominated date at interview.
4. An obstetric hospital was randomly chosen in proportion to the obstetric hospital of birth of SUDI cases over the previous four years.
5. Ethnicity was randomly allocated to each control in proportion to the ethnicities of the cases over the previous four years.
6. Random numbers were used to select a particular ethnic specific infant from among those born on the nominated date at that obstetric hospital. For obstetric hospitals where there were no deliveries of ethnic-specific babies on the nominated date, a randomly allocated direction indicator was used to indicate whether to go forwards or backwards in time to select an infant.

This selection meant that the distributions of the cases and the controls were very similar (over hospital, ethnicity and age) but there is no direct matching. The advantage of an unmatched study is that there will be no loss of efficiency because of failure to find a match. This method resulted in a control group that is enriched for the major risk factors (ethnicity and residence/socioeconomic status) and allows the identification of more subtle differences between cases and controls.

The initial plan was to select two controls for the anticipated number of cases, however, the participation rate of controls was lower than expected, so if the selected control could not be obtained, then a further control was selected. In total, 649 controls were selected.

Data collection for controls

The parents of control infants were sent a patient information sheet, and were phoned one to two weeks later to arrange an interview close to the nominated date. Written consent was obtained and the parents or guardians were interviewed and a "sleep scene" investigation conducted, the components of which were similar to the death scene investigation of the cases.

Variables

Most of the information for this report came from interviews with the parent or guardian.

Maternal ethnicity was self-reported. If missing it was taken from other sources, such as the notification information from NIIO. Multiple ethnicities could be given, and were prioritised using the following hierarchy: Māori, Pacific, Other and New Zealand European.[20] Bed sharing was

defined as sleeping on the same surface at the time of death or end of nominated sleep for controls. Maternal age (years), birthweight (kg) and age of infant (weeks) were treated as continuous variables. All other variables were categorised: sex (boy/girl), multiple birth (yes/no), number of previous live births (0, 1, 2, 3+), marital status (married, cohabitating, single), maternal smoking in pregnancy (yes/no), ever been breastfed (yes/no), position placed to sleep for the last sleep prior to death/nominated sleep (back, side, front) and sharing parental bedroom at the time of death/nominated sleep (yes/no; note: this refers to the parental bedroom, so an infant can be bed sharing but not be in the parental bedroom, such as sleeping on a sofa in the lounge).

Statistical analysis

Statistical analysis was carried out using the standard methods of the Mantel-Haenszel odds ratio analysis used in case-control studies.[21] Logistic regression for unmatched analysis of categorical variables was used to adjust for potential confounders. The multivariable analysis adjusted for: maternal ethnicity, maternal age, marital status, number of previous live births, age of infant, sex of baby, birth weight, singleton/multiple birth, breastfeeding, position placed to sleep, smoking in pregnancy, sharing parental bedroom and bed sharing. The interaction between maternal smoking and bed sharing was also examined. The analyses were conducted in SAS (Version 9.3, SAS Institute, Cay, NC, USA). Population attributable risk (PAR) for smoking, bed sharing and not sleeping in the parental bedroom were calculated to estimate the proportion of deaths explained by exposure to particular risk factors.[22] The number of SUDI cases not exposed to maternal smoking, bed sharing and not sharing the parental bedroom was calculated. Missing values were not imputed. Statistical significance was set at 5% level.

Ethics

Ethical approval for this study was obtained from Central Regional Ethics Committee (CEN/11/09/045). Parents/guardians of both cases and controls gave written consent.

Results

During the three-year study there were 303 deaths referred to the coroner that were considered for inclusion in the study. Excluded infants were:

- Born and domiciled outside New Zealand, n=1
- Greater than 12 months of age, n=43
- Less than seven days of age, n=63
- Non-accidental injury, obvious accidental causes, concealed pregnancies, n=19
- No autopsy, n=1
- Perinatal asphyxia, perinatal problems, including complications of prematurity, n=8
- Identified cause with prodromal symptoms and signs, n=18
- Congenital anomalies that clearly led to death directly, n=13

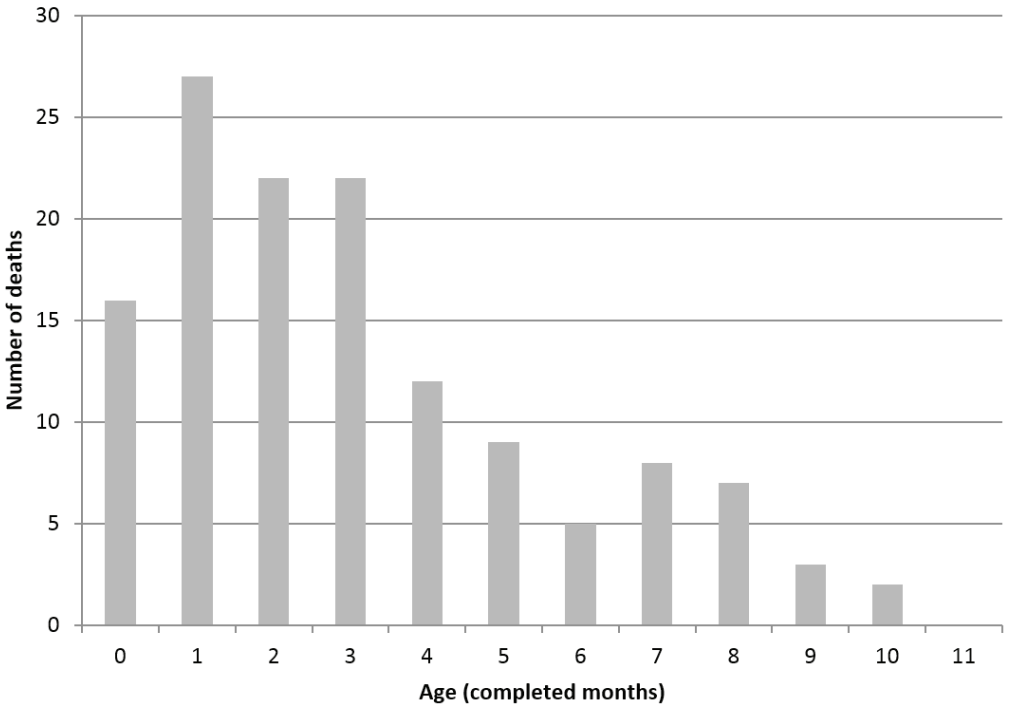
Thus there were 137 SUDI cases. These deaths were subcategorised as:

- Clear asphyxia mechanism, n=20
- Unsafe sleeping, n=50 and an additional 18 which also had minor pathological findings not thought to have contributed to the death
- Presence of minor pathological findings not thought to have contributed to the death, n=13 and
- Unexplained, n=36.

The SUDI mortality rate in the study period was 0.76/1,000 live births. The rate for Māori was 1.46/1,000, Pacific 1.01/1,000 and non-Māori non-Pacific (predominantly European) 0.45/1,000. The SUDI mortality rate by region was Upper North Island 0.70/1,000, Central North Island 1.00/1,000, Lower North Island 0.75/1,000 and South Island 0.61/1,000. There was no seasonal distribution of SUDI cases (spring=26, summer=38, autumn=31, winter=38). Figure 1 shows the age distribution of cases. The peak occurrence is 1–3 months of age with 74% of all SUDI deaths occurring before four months of age.

Parental (or guardian) interviews were completed in 133 (97%) cases and 258 (40%) controls. The initial interview occurred at

Figure 1: The age distribution of SUDI cases.



Number of deaths in the first month of life includes deaths from seven days of age through to 27 days of life.

a median of six days (interquartile range two to 12 days). No information other than ethnicity and obstetric hospital of birth were available for the non-participating controls.

Tables show the univariable and multivariable odds ratios (OR) for the variables relating to sociodemography, pregnancy, infant and infant care practices (Table 1).

Ethnicity and age were included in the multivariable analysis as they were part of the selection criteria for the control population. Because there were missing values for some variables the final multivariable model had 99 cases and 255 controls.

Significant findings at the 5% level in the multivariable analysis were number of previous live births, maternal smoking in pregnancy, multiple births, position placed to sleep, bed sharing and the protective effect of sharing the parental bedroom.

Maternal smoking in pregnancy increased the risk of SUDI (adjusted OR=6.01, 95% CI=2.97, 12.15) and was present in 74% of cases. Infants placed prone (on their front) to sleep were at an increased risk (aOR=3.85, 95% CI=1.07, 13.89) compared to infants placed on their back to sleep. Infants placed on their side had a non-significant increased risk of SUDI (aOR=1.94, 95% CI=0.85, 4.43).

57.5% of deaths occurred in a bed sharing situation. Bed sharing increased the risk of SUDI (aOR=4.96, 95% CI=2.55, 9.64) and was present in 57.5% of cases. Infants not sharing the parental bedroom were at increased risk of SUDI (aOR=2.77, 95% CI=1.45, 5.30).

The interaction between bed sharing and maternal smoking in pregnancy was examined (Table 2). Infants of mothers who smoked in pregnancy and were bed sharing were at a markedly increased risk of SUDI (aOR=32.8, 95% CI=11.2, 95.8) compared with infants not exposed to maternal smoking and bed sharing. The combination of bed sharing and smoking was associated with 48.0% of the deaths. Infants only exposed to maternal smoking in pregnancy and not bed sharing had a non-significant increased risk of SUDI (aOR=1.91, 95% CI=0.77, 4.72), and infants only exposed to bed sharing but not maternal smoking in pregnancy also had a non-significant increased risk (aOR=1.59, 95% CI=0.52, 4.87).

In the multivariable model, 34 cases and two controls were removed due to missing values. To assess the effect of this on the ORs, a multivariable model was run removing four variables with varying

Table 1: The number (percentage) or mean (SD) and univariable and multivariable odds ratios (95% CI) of sociodemographic, pregnancy, infant and infant care practice variables.

	Cases (%) N=133	Controls (%) N=258	Univariable OR (95% CI)	Multivariable* OR (95% CI)
Ethnicity (missing=6)			p=0.08	**p=0.048**
European	28 (22.0)	73 (28.3)	1.00	1.00
Māori	63 (49.6)	135 (52.3)	1.22 (0.72, 2.06)	0.57 (0.26, 1.26)
Pacific	19 (15.0)	34 (13.2)	1.46 (0.72, 2.97)	1.61 (0.58, 4.48)
Other	17 (13.4)	16 (6.2)	**2.77 (1.23, 6.23)**	2.15 (0.65, 7.12)
Marital status (missing=23)			**p=0.002**	p=0.62
Married	19 (17.0)	89 (34.8)	1.00	1.00
Cohabiting	53 (34.6)	100 (39.1)	**2.48 (1.37, 4.51)**	1.52 (0.65, 3.58)
Single	40 (35.7)	67 (26.2)	**2.80 (1.49, 5.26)**	1.46 (0.60, 3.57)
Number of previous live births (missing=13)			**p<0.0001**	**p=0.011**
0	63 (52.5)	59 (22.9)	1.00	1.00
1	14 (11.7)	62 (24.0)	**0.23 (0.11, 0.42)**	**0.23 (0.09, 0.57)**
2	16 (13.3)	41 (15.9)	**0.37 (0.19, 0.72)**	0.61 (0.24, 1.55)
3+	27 (22.5)	96 (37.2)	**0.26 (0.15, 0.46)**	**0.39 (0.17, 0.89)**
Maternal age at birth (mean years, SD) (missing=11)	25.3 (6.5)	28.7 (6.6)	**p<0.0001**	p=0.096 0.96 (0.91, 1.01)
Smoking during pregnancy (missing=9)			**p<0.0001**	**p<0.0001**
No	32 (25.8)	167 (64.7)	1.00	1.00
Yes	92 (74.2)	91 (35.3)	**5.28 (3.28, 8.50)**	**6.01 (2.97, 12.15)**
Multiple birth (missing n=5)			**p=0.010**	**p=0.029**
Yes	8 (6.3)	4 (1.6)	**4.23 (1.25, 14.34)**	**6.57 (1.21, 35.70**)
No	120 (93.8)	254 (98.4)	1.00	1.00
Baby sex (missing=0)			p=0.31	p=0.27
Female	56 (42.1)	95 (36.8)	1.00	1.00
Male	77 (57.9)	163 (63.2)	0.80 (0.52, 1.23)	0.71 (0.39, 1.31)
Birthweight (mean g, SD) (missing n=14)	3158 (619)	3466 (581)	**p<0.0001** **0.42 (0.28-0.61)**	p=0.057 0.60 (0.36, 1.01)
Age of infant (mean weeks, SD) (missing=0)	14.3 (18.1)	15.3 (10.4)	p=0.50	p=0.98 1.00 (0.97, 1.03)
Position placed to sleep (missing=7)			**p=0.0006**	**p=0.051**
Back	83 (65.9)	215 (83.3)	1.00	1.00
Side	31 (24.6)	31 (12.0)	**2.59 (1.48, 4.53)**	1.94 (0.85, 4.43)
Front	12 (9.5)	12 (4.7)	**2.59 (1.12, 6.00)**	**3.85 (1.07, 13.89)**
Breastfed (missing=5)			**p=0.014**	p=0.50
Yes	115 (89.8)	248 (96.1)	1.00	1.00
No	13 (10.2)	10 (3.9)	**2.80 (1.19, 6.58)**	1.53 (0.45, 5.24)
Sharing parental bedroom (missing=6)			**p=0.006**	**p=0.002**
Yes	69 (54.3)	177 (68.6)	1.00	1.00
No	58 (45.7)	81 (31.4)	**1.84 (1.19, 2.84)**	**2.77 (1.45, 5.30)**
Bed sharing (missing=6)			**p<0.0001**	**p<0.0001**
No	54 (42.5)	212 (82.2)	1.00	1.00
Yes	73 (57.5)	46 (17.8)	**6.23 (3.88, 10.02)**	**4.96 (2.55, 9.64)**

Bold indicates significant at the 5% level.
*Variables in model: ethnicity, marital status, number of previous live births, maternal age, maternal smoking in pregnancy, multiple birth, sex, birthweight, age of infant, position placed to sleep, breastfeeding, sharing parental bedroom and bed sharing.

Table 2: Interaction between maternal smoking in pregnancy and bed sharing on risk of SUDI.

		Cases	Controls	Univariable OR (95%CI)	Multivariable * OR (95%CI)
Smoking	Bed sharing	(missing=10)		**p=0.033** (interaction)	**p=0.002** (interaction)
No	No	21 (17.1)	138 (53.5)	1.00	1.00
No	Yes	11 (8.9)	29 (11.2)	**2.75 (1.17, 6.48)**	1.59 (0.52, 4.87)
Yes	No	32 (35.2)	74 (28.7)	**2.64 (1.33, 5.26)**	1.91 (0.77, 4.72)
Yes	Yes	59 (48.0)	17 (6.6)	**31.1 (14.0, 69.3)**	**32.8 (11.2, 95.8)**

Bold indicates significant at the 5% level.
*Bed sharing and maternal smoking combinations were adjusted for ethnicity, marital status, number of previous live births, maternal age, maternal smoking in pregnancy, multiple birth, sex, birthweight, age of infant, position placed to sleep, breastfeeding and sharing parental bedroom.

amount of missing data (marital status, 23 missing; parity, 13 missing; maternal age, 11 missing; and birthweight, 14 missing). This showed an increase in the point estimates of the odds ratios (smoking in pregnancy OR=6.44, 95% CI=3.52, 11.81; not sharing parental bedroom OR=2.98, 95% CI=1.66, 5.36; bed sharing OR=6.27, 95% CI=3.48, 11.30). Additionally we compared the prevalence of the four major risk factors for those cases not able to be included in the multivariable model (n=34) and those in the multivariable model. Those not in the model had a higher prevalence of all risk factors and were significantly more likely to be prone or side sleepers and to not ever breastfeed (data not shown).

Population attributable risk (PAR) for maternal smoking in pregnancy was 60%, bed sharing 48% and infants not sleeping in parental bedroom 31% for this high-risk population. If a representative control population was selected the odds ratios would have been higher but the prevalence lower. However, this results in a similar PAR (Table 3).

Discussion

The SUDI mortality rate in this three-year study was 0.76/1,000 live births, and the rate was higher in Māori (1.46/1,000) than Pacific (1.01/1,000) and non-Māori non-Pacific (mainly European, 0.45/1,000). SUDI occurred more frequently in male infants than female as expected. Deaths were more common in twins and those that were low birthweight. The peak age of death was 1–3 months of age. The age distribution is slightly younger than in the New Zealand Cot Death Study, which is consistent with other recent population-based studies, such as the SWISS study in southwest England.[23] Younger infants are probably more vulnerable to the combined effects of maternal smoking in pregnancy and bed sharing, which results in an interaction between bed sharing and infant age as well as with maternal smoking.[24,25] Infants of young and not married mothers were at higher risk of SUDI in the univariable analysis but not after adjustment for potential confounders.

Table 3: Proportion of the population exposed to risk (p), relative risk (OR) and population attributable risk (PAR) seen in this study and the estimated p, OR and PAR if the controls had been representative of all births.

	High-risk controls			**Representative of all births**		
	p	**OR**	**PAR**	**p**	**OR**	**PAR**
Smoking	0.353	5.28	0.60	0.159*	15.20	0.69
Bed sharing	0.178	6.23	0.48	0.134†	8.74	0.51
Not sharing parental bedroom	0.314	1.84	0.21	0.304†	1.93	0.22

*From the New Zealand National Maternity Collection (PMMRC).
†Data from 2013 Auckland survey of infant care practices (Hutchison et al, 2015).

The major modifiable risk factor was maternal smoking in pregnancy. The mothers of 74% of cases smoked. Infants of smokers were at a six-fold increased risk of SUDI compared to infants of non-smokers. Maternal smoking is a well-established risk factor for SIDS. A meta-analysis found that the magnitude of the risk increased after the decrease in prone sleeping position.[26] The OR reported here is even higher than that reported previously. The population attributable risk (PAR) is 60% for this high-risk population. As we sampled high-risk controls the smoking rate was higher (35.3%) than that reported nationally in pregnancy (15.9%).[27] Thus the magnitude of the risk would have been even higher if we had compared the cases to a nationally representative sample of births (estimated OR=15.2). Furthermore, the PAR would have been 68% if all births are considered.

Consistent with previous retrospective reports,[15,16] 57.5% of infants died while bed sharing, compared with 17.8% of the high-risk controls bed sharing. Bed sharing increased the risk five-fold. The PAR is 48% in this higher-risk population (and 51% in all infants).

The original New Zealand Cot Death Study identified a significant interaction between maternal smoking and bed sharing,[3] which has been confirmed by other studies.[25,28] In this study, the risk of SUDI for an infant exposed to both these risks was strikingly high, a 32-fold increased risk, compared with infants not exposed to either risk factor. It should be noted that if an infant was only exposed to one of these risk factors (smoking only or bed sharing only) the risk was increased but did not reach statistical significance in the multivariable analysis (smoking only OR=1.91, bed sharing only OR=1.59). This should not be interpreted as meaning these risks, such as bed sharing in the absence of maternal smoking, are safe, as previous larger studies have identified these as a significant risk.[24,25] The absence of a statistically significant result is almost certainly a consequence of the small sample size and the fact that we chose high-risk controls, which meant they were more similar to the cases, as seen with maternal smoking.

The interaction between bed sharing and smoking has been shown in other studies to be further complicated by alcohol and drug use.[25] These factors have not been included in this preliminary report of the results of the present study, but will be examined in more detail in subsequent analyses and publications. The importance of the present study is to draw attention to the extremely high risk attached to bed sharing by mothers who smoke.

Despite these controls being high risk, the study showed that only 4.7% of control infants were placed prone to sleep and only 12.0% were placed on their side. Clearly the message "Back to Sleep" has been received and implemented in the majority of this population. However, the study also illustrates that continued promotion of this message is required as prone sleep position increased the risk 3.8-fold and side 1.9-fold. Although the side sleeping position did not reach statistical significance, the point estimate is consistent with meta-analyses, which show a two-fold increased risk.[29] In the period before "Back to Sleep" there was a large excess of winter deaths, and there was a north-south mortality gradient. Following the "Back to Sleep" campaign these risks were attenuated. Now that very few infants sleep prone these risks have been almost entirely eliminated.

Almost half (45.7%) of the cases were not sharing the parental bedroom, and this was associated with an increased risk of SUDI. This risk factor has been recognised since 1996.[7] In the original study we showed that the protective effect was from sharing with adults (plus/minus other children) but not with children only. The effect was separate to bed sharing, and the lowest risk was in infants that shared the parental bedroom but not the parental bed. More could be done to promote the protective effect of infants sharing the parental bedroom, as in this high-risk population 31% of infants did not share the parental bedroom and the population attributable risk was 21%.

The association with the number of previous live births were unexpected. 52.5% of cases were first born vs 20.4% in the New Zealand Cot Death Study, and the risk decreased with increasing parity, whereas the risk of SUDI associated with parity is usually reported to increase. For controls, 23% were born to primiparous mothers compared with the national figure of 41% in

2014. This might in part be due to selection of high-risk controls who tend to have more children than the general population or may be due to selection bias—controls are more likely to participate if they have had previous children.

It is worth examining some of the risk factors that did not reach statistical significance. Lack of breastfeeding was significantly associated with risk of SUDI in the univariable analysis but not after adjustment for potential confounders. Breastfeeding rates are high in New Zealand, and even in this high-risk control population only 10 (3.9%) control infants were not breastfed, thus limiting our ability to identify this as a risk. Our original study identified lack of breastfeeding as a risk,[1,2] and this has been confirmed in subsequent meta-analyses.[30] We should continue to promote breastfeeding for this and other infant and maternal health benefits. The mean birthweight of cases was 308g less than that of the controls. However, after adjustment for other factors this approaches significance (p=0.057).

Just 21 cases over three years were not exposed to smoking in pregnancy, bed sharing or front or side sleeping position, which illustrates how few SUDI deaths might occur if no baby was exposed to these risks.

The strengths of the study were an excellent participation rate by the cases (97%), and that only one potential case was excluded due to no autopsy. However, a number of limitations must be considered. Firstly, the number of SUDI deaths (n=137) was 35% lower than that expected (n=210). This is real as shown by the 29% reduction in the number and rate of postperinatal deaths that has been reported recently.[31] This reduction was attributed to the Safe Sleep Programme, which consists of universal education and targeted supply of Infant Safe Sleep Devices (wahakura and Pepi-Pods) to infants at high risk. The wahakura is a flax basket in which the infant sleeps that can be taken into the parental bed. The Pepi-Pods is a polypropylene version of the wahakura. In our study no deaths occurred in a wahakura or Pepi-Pods. This reduction in SUDI mortality of course limits the power of the study to detect differences between the cases and controls. Secondly, we chose high risk controls, who were more likely to be socioeconomically disadvantaged, Māori and smokers. Ethnic minorities and those socioeconomically disadvantaged are less likely to participate in surveys,[32] and this resulted in only 40% of the selected controls participating, which introduces the possibility of selection bias. However, the controls that did participate were still of high risk (Māori 52%, smokers 35%).

Three (1%) of the controls and 34 (25%) of the cases had missing variables, and thus were excluded from the multivariable models. Removing four variables with the most missing data increased the point estimates for maternal smoking in pregnancy, not sharing the parental bedroom and bed sharing. Further, those with missing data had a higher prevalence of all risk factors examined. This would suggest that the increase in the point estimates in the model excluding the four variables is likely a combination of the exclusion of these variables, which are related to socioeconomic disadvantage, and the inclusion of some higher risk cases. Thus our results are likely to be conservative.

In conclusion, this study has shown that many of the risk factors, which were identified in the original New Zealand Cot Death Study (1987–1989), are still relevant today. Our findings indicate that the prevention messages are still applicable today, indeed these findings suggest the prevention messages should be reinforced. If these identified risks could be avoided, there could be a further substantial fall in SUDI mortality to just seven infant deaths per annum.

Competing interests:
Nil.

Acknowledgements:
We thank the Health Research Council of New Zealand for funding the feasibility study and this study, Cure Kids for their support of EAM and JMDT, and Communio who managed the project. We also thank Yvonne Ledesma-Allard, Lead Medicolegal Investigator, Miami-Dade Medical Examiner, Miami, Florida who trained the SUDI Liaison staff in conducting scene investigations.

A steering group met monthly by teleconference. This comprised: Professor Ed Mitchell (Principal investigator and Chair), Chief Coroner Neil MacLean, Ms Jackie Andrews (Office of the Chief Coroner), Coroner Morag McDowell, Mr Glenn Dobson (Charlotte Davies) (Operations Manager, Coronial Services), Mr Dave Aro (Director, Communio), Ms Melanie MacFarlane (project manager), Professor Dawn Elder (co-investigator), Dr Nick Baker (co-investigator and chair of the Child and Youth Mortality Review Committee), Associate Professor Beverley Lawton (Māori adviser), Dr Jane Vuletic and Dr Jane Zuccollo (pathologists), Inspector Patricia O'Shaughnessy (NZ Police).

We thank the SUDI Liaison staff who conducted the interviews: Shelley Jonas, Elaine McLardy, Genevieve Ali, Jazz Heer, Tracy Rewiri, Rebecca Passi and Judy McIntyre.

Finally, we especially thank the families of both bereaved and control infants for participating in this study, which would not have been possible without their willingness to share their stories with us.

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URL:
http://www.nzma.org.nz/journal/read-the-journal/all-issues/2010-2019/2017/vol-130-no-1456-2-june-2017/7262

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